



## Original Article

# The development and psychometric assessment of a questionnaire to assess sleep and daily troubles in parents of children and young adults with severe psychomotor impairment



Anna L. Tietze<sup>a</sup>, Boris Zernikow<sup>a,\*</sup>, Michael Otto<sup>a</sup>, Gerrit Hirschfeld<sup>a</sup>, Erik Michel<sup>b</sup>, Michelle Koh<sup>c</sup>, Markus Blankenburg<sup>a</sup>

<sup>a</sup> Chair for Children's Pain Therapy and Paediatric Palliative Care, Witten/Herdecke University, Germany

<sup>b</sup> Kinderklinik, Klinikum Friedrichshafen, Friedrichshafen, Germany

<sup>c</sup> Great Ormond Street Hospital for Children NHS Trust, London, UK

## ARTICLE INFO

## Article history:

Received 30 March 2013

Received in revised form 7 September 2013

Accepted 10 September 2013

Available online 20 November 2013

## Keywords:

Screening

Sleep disturbances

Children with severe psychomotor impairment

Parents' impairment

Parents' questionnaire

Validity

HOST

## ABSTRACT

**Background:** Children with severe psychomotor impairment (SPMI) often experience sleep disturbances that severely distress both the child and his or her parents. Validated questionnaires for the assessment of parents' distress related to their child's sleep disturbances are lacking.

**Methods:** We developed and validated a new questionnaire, the HOST (holistic assessment of sleep and daily troubles in parents of children with SPMI) to assess the effect of the sleep disturbances in children with SPMI on their parents. The questionnaire was developed based on published data and expert opinion, and it was refined via direct consultation with affected parents. Its psychometric characteristics were assessed in a sample of parents of 214 children with SPMI. It was retested using a random subsample of the participants.

**Results:** Explorative factor analysis revealed that the HOST was composed of four scales. Fit indices, item analysis, and convergent validity (coherence with preexisting instruments of sleep disturbances and health status) were adequate. Retest analysis ( $n = 62$ ) revealed high stability of the HOST questionnaire and adequate replication validity.

**Conclusion:** Sleep-related difficulties significantly impact the sociomedical characteristics of the parents of children with complex neurologic diseases. Typically, parents are severely affected in various aspects of daily life (i.e., medical health, social life, professional life). The HOST proved to be a valid, reliable and economical assessment tool of sleep-related difficulties in parents and relatives of children with SPMI. The HOST is capable of identifying individuals and specific areas requiring intervention.

© 2013 Elsevier B.V. All rights reserved.

## 1. Introduction

A unique feature of pediatric populations is that the health and well-being of parents can be strongly affected by the medical condition of their children. This statement is especially true in highly disabling pediatric conditions such as severe psychomotor impairment (SPMI). SPMI is associated with multiple diseases, including childhood neuromuscular disorders (e.g., muscular dystrophies, spinal muscular atrophies), cerebral palsy, and neurodegenerative and metabolic diseases (e.g., mucopolysaccharidosis, neuronal ceroid lipofuscinosis). These children are impacted by both

intellectual impairments and physical disabilities. The occurrence of SPMI in children and adolescents is 1–3% [1,2]. This condition most often results from perinatal insults (i.e., prematurity, asphyxia), syndromes, and other genetic or metabolic diseases [3]. Approximately 50–80% of these children experience sleep disturbances (i.e., delayed sleep onset, disturbed sleep–wake cycles, difficulties in waking [4,5]), which result in restlessness and hyperactivity during the day [6,7]. There are various factors inherent to the underlying conditions provoking sleep disturbances in children with SPMI. On the one hand, there are several physical disabilities, including children's impaired ability to reposition their bodies, combined sensory and perceptual impairment, reduced natural melatonin secretion, or epilepsy which may contribute to SPMI. However, on the other hand, disturbed and altered cognitive processes (e.g., mental, communication, behavioral impairment, overexcitation) that affect sleep may trigger sleep disturbances [4].

\* Corresponding author. Address: Chair for Children's Pain Therapy and Paediatric Palliative Care, Witten/Herdecke University, Children's Hospital Datteln, Dr. Friedrich Steiner Str. 5, D-45711 Datteln, Germany. Tel.: +49 2363 975 187; fax: +49 2363 975 181.

E-mail address: [B.Zernikow@kinderklinik-datteln.de](mailto:B.Zernikow@kinderklinik-datteln.de) (B. Zernikow).

Sleep disturbances not only affect children with SPMI but also the parents of the children, which represents a highly relevant medical and social problem [6,8–10]. The majority of caregivers report that their own sleep quality is poor, which may cause fatigue, sleep problems, somatic complaints, depression, and other psychiatric disorders such as anxiety disorders or affective disturbances [11–13]. In one study, 74% of the parents of children with SPMI reported that their quality of life (QoL) or that of the child's healthy siblings was moderately to severely affected by the care needs or restrictions in everyday life related to the child's disease [14]. In a recent study by Wayte et al. [15], 40% of the mothers of children with cerebral palsy reported poor sleep quality; of whom, 44% endorsed depressed mood. Pediatric and maternal sleep disturbances were significantly correlated. In contrast to children with cerebral palsy, children with SPMI are unable express their sleep problems. Another study investigated sleep-related troubles in children with intellectual disabilities and their parents [16]. The author's report higher levels of maternal stress and depression and lower maternal well-being in mothers of children with autism compared to mothers with normally developed children [16]. It is important to note that these children do not have physical impairments.

Children with SPMI experience physical disabilities and cognitive impairments, which likely result in increased parental concerns and troubles with pediatric sleep and nighttime care. There is little information available regarding sleep disturbances in the parents of children with SPMI. Furthermore, the conceivable consequences of daytime sleepiness, social life, and family functioning remain unclear.

The direct effect of sleep disturbances in pediatric SPMI on the sleep patterns and daily troubles of the affected children's caregivers has not been well-studied [11,12,17,18]. Objective and efficient measures of sleep disturbances and daily troubles in parents with handicapped children are lacking and are urgently needed. The validity of questionnaires used in published studies that assess parental burden generally is low. The investigators of these studies adapted preexisting pediatric assessments to their parent samples; however, there was no explicit reevaluation or recalibration of these instruments [19]. The use of these tools to assess sleep disturbances and daily troubles is not meaningful; they were developed for use in healthy populations (families and parents without handicapped children). However, these tools do not necessarily reflect the effect that a child's disease has on the sleep patterns of the parents (i.e., the necessity of permanent caregiving and the sorrows and fears of the parents). To assist clinicians confronted with family sleep problems and enable targeted intervention, it is important to assess the effect of the sleep problems in children with SPMI and the worries of their parents regarding the children's diagnoses and development on each of the parents' functioning. In particular, there is no instrument that addresses sleep and the problems in everyday life that result from sleep disturbances to date (i.e., physical and mental functioning, partnership, social contacts, effect on job performance).

The aim of our report was to assess sleeping patterns and the QoL in parents of children with SPMI. We developed and validated an assessment tool for this purpose, called the HOST (*holistic assessment of sleep and daily troubles in parents with children with SPMI*) questionnaire.

## 2. Methods

We conducted a multicenter, questionnaire-based, cross-sectional survey for parents of inpatients and outpatients with severe cognitive and psychomotor impairments due to severe neurologic or metabolic disease or related to a congenital or genetic syndrome.

### 2.1. Patient recruitment

Parents and patients were recruited from three inpatient institutions (Children's Hospital Datteln, Datteln, Germany; Short-term Home "Kleine Oase Datteln"; and Children's Hospice Balthasar in Olpe, Germany) and one outpatient facility (Paediatric Palliative Care Team Datteln, Germany). Inclusion criteria were as follows: (1) patients presenting at one of the listed facilities between July 2008 and January 2010, (2) children or adolescents diagnosed with SPMI, (3) age ranging from neonate to young adulthood (zero days–25 years), and (4) parents or caregivers who are able to adequately communicate in German either verbally or in writing.

All of the patients who satisfied the inclusion criteria were contacted by phone and informed of the aims of our study. If the patient was eligible and the parents consented to participate in the study, they were sent a set of questionnaires, a letter of intent, a consent form, and a prepaid return envelope. All parents who did not return the questionnaires after 2 weeks were contacted via phone by the study coordinator (ALT). Parents who did not respond to the phone call did not participate in the study.

### 2.2. Introducing a new assessment tool: the HOST questionnaire

The HOST questionnaire was designed to evaluate the effects experienced by the parents or caregivers of children with SPMI-related sleep disturbances. The basic concept and structure were adapted from the Pittsburgh Sleep Quality Index (PSQI), an internationally validated assessment tool recommended by the German Sleep Society to assess adults with sleep disturbances. For use in the parents of children with SPMI, we modified the test items to reflect the specific life restrictions and mental or somatic distress experienced by affected parents and the effect of their child's disease on parents' sleep (i.e., due to the permanent care needs of the child and the sorrows, worries, and fears of the parents).

The HOST is meant to be completed by parents, answering questions regarding their own sleep problems and the disruption of their lives resulting from their child's sleep problems during the preceding 4 weeks. In the HOST, the construct (disturbed) sleep addresses this family problem using a biopsychosocial model. In contrast to the PSQI, the HOST questionnaire allows for the specific assessment of sleep-related impairments in parents of a handicapped child. The HOST also allows the parents' nighttime disruption due to the amount of time required to care for their child to be mapped. In addition, the HOST asks for data regarding family changes and a variety of problems (i.e., partnership, social contacts, effect on job performance) resulting from the sleep disturbance of the parent or the child. This information is essential to appropriately treat and support these clients.

The HOST was developed in four stages: (1) a comprehensive literature review provided an overview of the current state of research and comprehensive standards (i.e., existing tools such as the PSQI), which resulted in the first draft of the HOST; (2) interviews with three pediatric experts on sleep and sleep disorders, which resulted in the second draft of the HOST; (3) interviews with 20 parents of children with SPMI and sleep disturbances (pretest) to test the second draft of the HOST for understandability, significance of meaning, clarity, and feasibility, which resulted in a preliminary final version of the HOST; and (4) an analysis of the pretest HOST data, which resulted in the final version of the HOST.

#### 2.2.1. Literature review

The literature review has been previously published [4]. It demonstrated that sleep disturbances in children with SPMI are a significant clinical problem for the affected child and his or her parents and family. It is obvious that a suitable and validated questionnaire for the assessment of the disruption in parents' lives

resulting from their child's sleep problems is urgently needed. From these data, we designed the first draft of the HOST, which consisted of 85 items.

### 2.2.2. Expert opinions

We identified three experts on pediatric sleep (Bernhard Schlüter [BS, Children's Hospital Datteln, Witten/Herdecke University Germany], Alfred Wiater [AW, Children's Hospital Porz am Rhein, Germany], and Stefan Eitner [SE, Hospital of Paediatrics and Youth Medicine, University Medical Center Tübingen, Germany]). In daily practice, these experts engage with parents whose sleep and overall lives are disrupted by their children's sleep difficulties; we asked these individuals to scrutinize the first draft of the HOST questionnaire.

The three experts focused on the following aspects: (1) in the HOST, the broad domains of the effect of the child's sleep disturbance on the parents should be clearly specified; (2) the proposed domains of the scale must be relevant and comprehensive with regard to the parents of a child with SPMI; (3) the selected domains should be mutually exclusive to prevent ambiguity in scoring, administration, and interpretation; and (4) any difficulties and problems described in the HOST should be written in terms that are understandable and relevant to the parents of a child with SPMI. The first draft of the HOST was modified according to the experts' suggestions, resulting in the second draft of the HOST, which consisted of 70 items.

### 2.2.3. Pretest parents' interviews

We interviewed a sample of 20 parents presenting with their children to the Department of Neuropaediatrics, Children's Hospital, Datteln. These parents had children with SPMI who experienced sleep disturbances. The parents were asked to complete the second draft of the HOST and to assess its form, wording, and order of the questions. The aim of the pretest was to determine if the questions were understandable and enabled respondents to answer them to the best of their ability.

Overall, the parents positively judged the second draft of the HOST questionnaire. Ambiguous items defined as those items that more than 20% of the respondents judged as difficult to understand were revised by the authors and reassessed by the parents. Redundant items, those that more than 20% of the respondents judged as redundant, were removed.

### 2.2.4. Analysis of pretest HOST data

Through analysis of pretest data, items were identified as irrelevant (8 items), redundant (5 items), or insignificant by parents' opinions (3 items). Together these 16 items were removed, resulting in the final version of the HOST, which consists of 54 items. The final HOST questionnaire consists of two parts (Table 1). There are some descriptive questions regarding the family's living situation, and 16 items are aimed to assess parental sleep-related difficulties in various areas of sleep, physical and mental functioning, social functioning, and occupational functioning. These items reflect the burden factors on the patient's relatives.

Part one (38 items) of the HOST gathers information regarding general sociodemographic aspects of the parents, their child, and parental sleep components (sleep environment, sleep conditions, sleep onset latency, duration of sleep, sleep efficiency) (Table 1). Sleep latency is defined as the length of time required to accomplish the transition from full wakefulness to sleep. A time span of up to 30 min is considered normal [20]. Sleep efficiency is defined as the ratio of time spent asleep (total sleep time) to the amount of time spent in bed. Sleep efficiency of good sleepers generally is greater than 85%. An efficiency less than 65% is considered low [20]. Sleep duration was assessed. Of their actual sleep, parents were asked: "how many hours of sleep did you actually get during

the night (this may be different than the number of hours you spend in bed)?"

These measures estimate the parents' sleep parameters, which include subjective self-report measures rather than a detailed objective record of sleep parameters only. Part 2 (16 items) gathers information regarding the frequency of sleep disturbance and the associated consequences of the lack of sleep (sleep disturbances, impairments in physical/mental functioning, impairments in social functioning, and impairments in occupational functioning) (Table 1). These items were analyzed to yield a disturbance score. Each item is scored using a 5-point scale (1 = never; 2 = less than once a week/rarely; 3 = once or twice a week/sometimes; 4 = three or four times a week/frequently; 5 = five or more times a week/always).

### 2.2.5. Translation into English

The HOST is intended for international use. We translated the German questionnaire into English using one of the most common translation techniques in international research (back translation) [21]. Back translation stipulates that the questionnaire is to be translated by one translator from the source language into a target language. A second translator, who is unaware of the source language, translates the questionnaire back into the source language. Both translators were bilingual native speakers. The original and the retranslated versions are then scrutinized for differences. The back translation process was repeated twice to avoid any errors and misinterpretations [21]. The German version was used in our study.

### 2.2.6. Retesting of HOST

Retesting was based on the data from a simple random sample ( $n = 62$ ) of parents of the study population. The sample was identified using the random sampling procedure in the SPSS statistical software package (IBM SPSS Statistics 21, IBM Corporation Germany, Ehningen, Germany). All parents who were selected for retesting agreed to participate; retesting was identically performed to the original testing, but after 8 weeks had passed. Retesting included an additional interview of the parents regarding the questionnaire. The interview directly inquires if there were any changes in family life not assessed by the questionnaire and if parents have further questions, remarks, or suggestions regarding the item selection.

## 2.3. Reference questionnaires

To evaluate the HOST questionnaire, parents completed a small number of reference questionnaires to assess sleep quality and the sleep disturbance-related burden and effects on their QoL in addition to the HOST.

### 2.3.1. PSQI

The PSQI evaluates the frequency of sleep-disturbing events, sleep quality, habitual sleep efficiency, sleep latency, sleep duration, use of sleep medication, and daytime dysfunction over the previous 4 weeks. It consists of 18 quantitative items, which are assigned to seven components; the score of each component ranges from 0 to 3. The total score is the sum of the scores of the seven components and may range from 0 to 21. Higher total scores indicated worse sleep quality. The empirical cutoff value of five divides good sleepers and bad sleepers ( $>5$ ). The stability of the PSQI was assessed in three studies and was proven to be satisfactory with retest reliability ( $r_{tt}$ ) of 0.82–0.89 for the total score. The  $r_{tt}$  of the individual components was between 0.45 (daytime sleepiness, [22]) and 0.84 (sleep latency, [23]). In two studies, Cronbach  $\alpha$  for the component score of sleep disturbance ranged from 0.70 to 0.78 [24,25]. Cronbach  $\alpha$  for the total score was 0.77 [25]. The

**Table 1**

Structure of the HOST (holistic assessment of sleep and daily troubles in children with severe psychomotor impairments) questionnaire.

Components	No. of items
(1) Sociodemographic aspects	18
(a) General information on child	
(b) General information on family	
(c) General information on caregiver	
(2) Sleep	10
(a) Sleep conditions (locality; sleep disturbing factors)	
(b) Sleep onset latency	
(c) Duration of sleep	
(d) Sleep efficiency	
(3) Parental burdens and consequences of the child's sleep deficiency	16
(1) Sleep disturbances	
(2) Impairments of physical or mental functioning	
(3) Impairments of social functioning	
(4) Impairments of occupational functioning	

Abbreviation: No., number.

Components 1–4: information regarding general sociodemographic characteristics of the parents, their child, and parental sleep components.

Component 5: items are calculated to yield a burden score.

sensitivity of the total score was >80% in several samples of patients with sleep disturbances, and the specificity was in a similar range (83–87% [23,26]). We used the parental PSQI scores to describe the parent sample and to calculate the convergent validity of the HOST.

#### 2.3.2. Short-Form 36-item Health Survey

The Short-Form 36-item Health Survey (SF-36) is a disease-independent tool for the assessment of health-related QoL in patients. The SF-36 measures eight dimensions: physical functioning, role-physical, bodily pain, general health, vitality, scale functioning, role-emotional, and mental health spanning the two domains physical health or mental health. The internal consistency (Cronbach  $\alpha$ ) of the SF-36 subscales ranges from 0.57 to 0.94. The convergent and discriminate validity and sensitivity of the questionnaire has been determined to be stable and acceptable [27]. We used the SF-36 scores to describe the parent sample and calculate the convergent validity of the HOST.

#### 2.3.3. Epworth Sleepiness Scale

The Epworth Sleepiness Scale (ESS) is a widely used self-assessment tool for the measurement of daytime sleepiness in eight different everyday situations. Responses are indicated on a 4-point Likert scale (0 = no chance of dozing; 3 = high chance of dozing) to estimate the probability of dozing in several real-life situations. The total score ranges from 0 to 24. In healthy individuals, retest reliability of the ESS was appropriate after 5 months ( $r = 0.82$ ;  $P < .001$ ) [28]. The internal consistency of the ESS is high (Cronbach  $\alpha$ , 0.88 [28]). The ESS scores were used to describe the parent sample and to calculate the convergent validity of the HOST.

#### 2.4. Data analysis

We investigated the convergent validity by examining the interrelationships (single correlations) between the parental sleep-related burden as assessed by the HOST and the PSQI [26]. Further support of the construct validity of the HOST was assessed by correlating the HOST scales with thematically similar subscales of the SF-36 [27] and the ESS [28]. We estimated the stability of the HOST (8-week interval) via correlation and  $t$  tests in a simple random sample. Random sampling was conducted using SPSS. The reference values used to facilitate interpretation of individual results ( $t$  value, percentage rank, among others) were established. This study was approved by the Ethics Committee of Witten/Herdecke University, Germany.

### 3. Results

#### 3.1. Recruitment

##### 3.1.1. Inpatients

Between July 2008 and January 2010, 263 patients presented to the Children's Hospital, Datteln, and satisfied the inclusion criteria. The families consented to participate in the study by phone. Of these families, 57% returned the questionnaire. Eleven questionnaires were excluded from the analysis because more than 50% of the values were missing. The inpatient sample size was 139 individuals. The number of inpatient participants from other institutions was 58 individuals (Inpatient Children's Hospice 'Balthasar', Olpe,  $n = 15$ ; Short-term Home 'Kleine Oase', Datteln,  $n = 43$ ).

##### 3.1.2. Outpatients

There were 27 outpatient participants (Paediatric Palliative Care Team Datteln). A total of 50 families had been contacted and the response rate was 54%.

Data from 224 (60%) participants were analyzed. No intergroup differences in setting (inpatient or outpatient) were found (data not shown).

##### 3.1.3. Demographic characteristics

Of the children severely affected by psychomotor impairment, 25% had a neuromuscular disorder, 11% had a neurodegenerative disease, 13% had a specific syndrome, and 16% had a global developmental impairment of unknown cause (Table 2A). At the time of the survey, 80% of the patients had used their compulsory German nursing care insurance (see Table 2B for definition). A total of 130 children needed 24-h daily assistance with personal hygiene, feeding, or mobility (Table 2B). Detailed patient characteristics are published elsewhere [5].

The mothers' mean ages were 41.3 years ( $n = 212$ ; standard deviation [SD], 7.6 years). The fathers' mean ages were 43.8 years ( $n = 193$ ; SD, 7.3 years). Of the questionnaires, 95% were completed by the mother or foster mother and the remaining 5% were completed by the father or foster father. In contrast to mothers, most fathers were employed fulltime (Table 2B).

#### 3.2. Exploratory factor analysis

An exploratory factor analysis revealed a 4-factor structure with an explained variance of 76%. A scree plot analysis supported the 4-factor solution (Fig. 1). In the first step, the promax rotated factor



**Table 2A**  
Distribution of diagnoses (grouped).

	Primary diagnosis		Secondary diagnosis	
	<i>n</i>	%	<i>n</i>	%
Metabolic disorder/neurodegenerative disorder	23	11	–	–
Cerebral palsy/neuromuscular disorder	54	25	7	3
Cerebral ischemia	7	3	1	0
Cerebral hemorrhage	5	2	6	3
Cerebral inflammation	5	2	2	1
Brain tumor	2	1	2	1
Global developmental retardation (several causes)	35	16	3	1
Hypoxia	5	2	7	3
Traumatic brain injury	1	0	1	0
Severe spina bifida	5	2	2	1
Syndrome	28	13	1	0
Cerebral malformation	8	4	2	1
Other	4	2	3	1
Diagnosis not established	33	15	177	83
Total	214	100		

Abbreviation: *n*, number of patients.**Table 2B**  
Family characteristics.

<i>Children/young adults (n = 214)</i>	
Girls (%)	46.5
Age in y (range)	10.4 (0.1–25.0)
Age group	
<1	2
2–5	46
6–12	93
13–18	54
>18	19
Nursing level (%)	
None	12
(1) Requiring help at least once per day	8
(2) Requiring help at least three times per day	14
(3) Requiring around the clock care daily	58
<i>Parents (n = 214)</i>	
Nationality	
Single parents	79.9
German (%)	88.8
Other (Polish, Russian, other) (%)	11.2
Employment status	
Fathers (n = 176)	
Fulltime (%)	82.9
Part time (%)	4.5
Unemployed (%)	12.5
Mothers (n = 171)	
Fulltime (%)	11.1
Part time (%)	22.8
Unemployed (%)	66.1

Abbreviation: y, years.

Nursing level, adopted from the Nursing Care Statistics of the Federal Statistical Office.

(1) Individuals requiring help at least once per day with bodily care, food, or mobility in at least two activities in one or more domains. On average, care must require at least 90 min more per day than the amount of time needed to care for a healthy child of the same age.

(2) Individuals requiring help at least three times per day at different times of day with bodily care, food, or mobility. On average, care must require at least 3 h per day more per day than the amount of time needed to care for a healthy child of the same age.

(3) Individuals requiring daily help around the clock, including the night, in bodily care, food, or mobility. On average, care must require at least 5 h per day and the nurse must be on call at all hours.

structure was systematically examined. The items of the questionnaire were assigned respective scales to elucidate the factors resulting from the exploratory analysis. An item was regarded as correctly assigned if its primary factor loading was >0.50 and there were no cross-loadings >0.30 to another factor. Two of the items

were left in the solution due to contextual considerations, though the primary loading was <0.50 (How often did you have trouble sleeping because you worried about your child's condition in the future?) or the cross loading was >0.30 (Have your sleep interruptions due to your child's condition caused insufficient sleep?). Significant correlations between the scales were observed (each  $P < .01$ ; Table 4). The four scales provide an underlying common or global concept (moderate correlations), whereas each scale exhibits its own distinct contextual aspects. The results of reliability and item analysis (corrected item-total correlation, means of the various scales, and their respective SDs) are listed in Table 3.

### 3.3. Internal consistency, selectivity and difficulty

The internal consistency of the subscales (Cronbach  $\alpha$ ) and the range of corrected selectivity ( $r_{it}$ ) and item difficulty ( $P$ ) for each subscale are given in Table 3. The internal consistency of the subscales ranged between  $\alpha = 0.74$  (scale 4) and  $\alpha = 0.91$  (scale 2). For the small number of items, the internal consistency can be judged as excellent, reflecting the high capability of the HOST to serve as an assessment tool [29]. This finding is even more remarkable, considering that reliability is substantially dependent on test length [29]. The items of each individual scale assess the same type of impairments. The corrected sensitivity is moderate to high, and their value indicates adequate item selection [30]. The item difficulties ranged between  $P = .44$  (item: have the interruptions of your sleep due to your child's condition caused trouble staying awake while driving, eating meals, or engaging in social activities?) and  $P = .90$  (item: during the last few years, how many times did the lack of sleep due to your child's condition and associated stress cause absence from work?). Generally, the distribution of difficulties was negatively skewed, which presumably reflected the subsample of parents who were highly burdened and sleep deprived as a result of their child's disease.

### 3.4. Validity

There were multiple significant positive correlations between the HOST subscales and the PSQI scales; they were especially strong for the first three HOST scales (sleep disturbances, impairments in physical and mental functioning, and impairments in social functioning). The correlation values ranged from  $r = 0.23$  to  $r = 0.67$ . The strongest correlation was observed between HOST scales 1 (sleep disturbances) and 2 (impairments in physical and mental functioning) and the PSQI. HOST scale 4 (impairments in

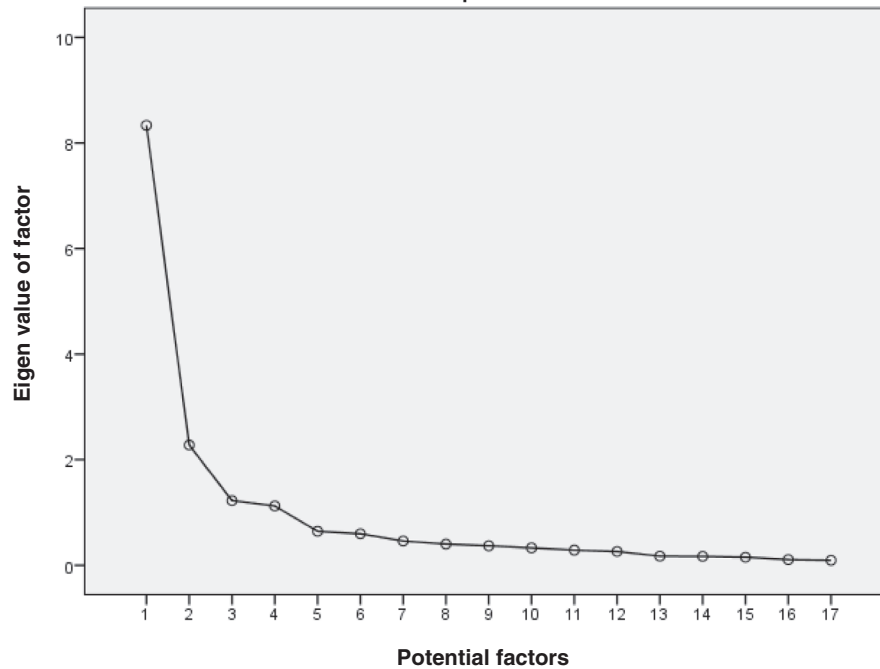


Fig. 1. Explorative factor analysis: scree plot.

Table 3

Characteristics of HOST (holistic assessment of sleep and daily troubles in children with severe psychomotor impairments).

Scale	M	SD	$\alpha$	$r_{it}$	P value	$r_{rt}$
S1: sleep disturbances	14.7	6.5	0.90			.78**
How often did you have trouble sleeping because you got up to look after your child?	3.1	1.5		.83	.62	
How often did you have trouble sleeping because you were woken up by your child?	3.2	1.5		.84	.64	
How often did you have trouble sleeping because you had to care for your child at night (e.g., reposition your child)?	2.8	1.6		.72	.56	
How often did you have trouble sleeping because you were worried that something could happen to your child while you were sleeping (e.g., seizure, spasm)?	2.4	1.6		.6	.48	
Have the interruptions of your sleep due to your child's condition caused insufficient sleep?	3.2	1.5		.78	.64	
S2: impairments of physical/mental functioning	12.8	6.0	0.91			.69**
Have the interruptions of your sleep due to your child's condition caused negative changes in mood?	2.4	1.4		.86	.48	
Have the interruptions of your sleep due to your child's condition caused increased bad temper or distress?	2.7	1.4		.86	.54	
Have the interruptions of your sleep due to your child's condition caused trouble staying awake while driving, eating meals, or engaging in social activities?	2.2	1.3		.74	.44	
Have the interruptions of your sleep due to your child's condition caused difficulties maintaining motivation to complete tasks?	2.9	1.5		.79	.58	
How often did you have trouble sleeping because you worried about your child's condition in the future?	2.6	1.5		.59	.52	
S3: impairments of social functioning	13.4	4.9	0.85			.88**
Over the years, how many times did lack of sleep due to your child's condition restrict your social activities (e.g., meeting friends)?	2.9	1.5		.5	.58	
Over the years, how many times did lack of sleep due to your child's condition have a negative impact on your relationship with your partner?	3.6	1.4		.82	.72	
Over the years, how many times did lack of sleep due to your child's condition have a negative impact on your affection and sex life with your partner?	3.2	1.5		.77	.64	
During the last few years, how many times did the lack of sleep due to your child's condition and associated stress cause a crisis in your partnership?	3.8	1.4		.67	.76	
S4: impairments of occupational functioning	8.4	2.0	0.74			.87**
During the last few years, how many times did the lack of sleep due to your child's condition and associated stress cause illness?	3.9	1.2		.6	.78	
During the last few years, how many times did the lack of sleep due to your child's condition and associated stress cause absence from work?	4.5	1.0		.6	.9	

Abbreviations: M, mean; SD, standard deviation;  $\alpha$ , Cronbach alpha (internal consistency);  $r_{it}$ , corrected item-total correlation;  $r_{rt}$ , test-retest correlation (retest sample,  $n = 62$ ).

P, item difficulty (the item difficulty index is one of the most useful and most frequently reported statistics obtained via item analysis. It is a measure of the proportion of examinees who correctly answered a specific item. The purpose of the difficulty index is to distinguish subjects with high expression of an assessed characteristic from subjects with lower levels of the characteristic).

\*\*  $P < .01$  (two tailed).

**Table 4**

Intercorrelation of the HOST (*holistic assessment of sleep and daily troubles in children with severe psychomotor impairments*) subscales (Pearson product moment correlation coefficients [*r*]).

	S2: impairments of physical and mental functioning	S3: impairments of social functioning	S4: impairments of occupational functioning
S1: sleep disturbances	.654**	.405**	.357**
S2: impairments of physical/mental functioning		.562**	.500**
S3: impairments of social functioning			.528**

\*\*  $P < .01$ .

**Table 5**

Convergent validity of HOST (*holistic assessment of sleep and daily troubles in children with severe psychomotor impairments*): correlation with the Pittsburgh Sleep Quality Index and the Epworth Sleepiness Scale (Pearson product moment correlation coefficients [*r*]).

Scale	HOST			
	S1: sleep disturbances	S2: impairments of physical/mental functioning	S3: impairments of social functioning	S4: impairments of occupational functioning
PSQI				
1. Subjective sleep quality	.55**	.64**	.48**	.27**
2. Sleep latency	.28**	.40**	.33**	.10 <sup>ns</sup>
3. Sleep duration	.49**	.44**	.33**	.13 <sup>ns</sup>
4. Habitual sleep efficiency	.52**	.47**	.35**	.22**
5. Sleep disturbances	.32**	.30**	.33**	.29**
6. Use of sleep medication	-.07 <sup>ns</sup>	.1 <sup>ns</sup>	.28**	.22**
7. Daytime dysfunction	.46**	.64**	.45**	.42**
Total score	.57**	.65**	.53**	.34**
Epworth Sleepiness Scale	.35**	.29**	.29**	.29**

Abbreviations: HOST, *holistic assessment of sleep and daily troubles in children with severe psychomotor impairments*; S, scale; ns, nonsignificant; PSQI, Pittsburgh Sleep Quality Index.

\*\*  $P < .01$  (2 tailed); ns, nonsignificant.

**Table 6**

Convergent validity of HOST (*holistic assessment of sleep and daily troubles in children with severe psychomotor impairments*) questionnaires: correlation with the Short-Form-36 (Pearson product moment correlation coefficients [*r*]).

Scale	HOST			
	S1: sleep disturbances	S2: impairments of physical/mental functioning	S3: impairments of social functioning	S4: impairments of occupational functioning
SF-36				
Physical functioning	-.32**	-.35**	-.28**	-.32**
Role-physical	-.35**	-.47**	-.33**	-.37**
Bodily pain	-.37**	-.44**	-.38**	-.30**
General health	-.25**	-.37**	-.39**	-.38**
Vitality	-.38**	-.64**	-.41**	-.34**
Social functioning	-.42**	-.57**	-.46**	-.39**
Role-emotional	-.32**	-.54**	-.41**	-.38**
Mental health	-.25**	-.53**	-.39**	-.29**

Abbreviations: HOST, *holistic assessment of sleep and daily troubles in children with severe psychomotor impairments*; SF-36, Short-Form 36-item Health Survey.

\*\*  $P < .01$  (2 tailed).

occupational functioning) least strongly correlated with the PSQI due to the lack of a contextual equivalent to HOST scale 4 on the PSQI (Table 5). All of the HOST scales adequately correlated ( $r = 0.54$ – $0.67$ ) with the scale total of the PSQI, supporting our contextual construct of sleep-related troubles or impairments and demonstrating that these instruments are not entirely equivalent but do map slightly different aspects. The HOST scales were significantly correlated with parental daytime sleepiness; HOST scale 2 (impairments in physical and mental functioning) exhibited the strongest correlation ( $r = 0.42$ ) (Table 5).

All of the HOST subscales were significantly correlated with the scales of the SF-36 ( $r = 0.25$  and  $-0.64$ ; Table 6). The highest

correlation existed between the HOST scale 2 (impairments in physical and mental functioning) and the scales vitality, social functioning and role-emotional of the SF-36. Another important result is that the HOST scales were not significantly correlated with children's age ( $r = -0.009$  to  $-0.2$ ).

### 3.5. HOST test–retest reliability

We evaluated the test–retest reliability of the HOST in a random subsample of 62 parents. The demographic and clinical characteristics of these parents were similar to those of the total sample (data not shown). We calculated Pearson product moment

**Table 7**HOST (holistic assessment of sleep and daily troubles in children with severe psychomotor impairments) conversion table for raw scores (*t* value distribution).

Raw score	S. 1 Sleep disturbances		S. 2 Impairments of physical/mental functioning		S. 3 Impairments of social functioning		S. 4 Impairments of occupational functioning	
	<i>t</i>	PR	<i>t</i>	PR	<i>t</i>	PR	<i>t</i>	PR
2							42	0.21
3							47	0.38
4							52	0.58
5	35	0.07	37	0.10	37	0.10	57	0.76
6	37	0.10	39	0.14	41	0.18	62	0.88
7	38	0.12	41	0.18	43	0.24	67	0.96
8	40	0.16	42	0.21	45	0.31	72	0.99
9	41	0.18	44	0.27	47	0.38	77	1.0
10	43	0.24	45	0.31	49	0.46	82	1.0
11	44	0.27	47	0.38	51	0.54		
12	46	0.34	49	0.46	53	0.62		
13	47	0.38	50	0.50	55	0.69		
14	49	0.46	52	0.58	57	0.76		
15	50	0.50	54	0.66	59	0.82		
16	52	0.58	55	0.69	61	0.86		
17	54	0.66	57	0.76	63	0.90		
18	55	0.69	59	0.82	65	0.93		
19	57	0.76	60	0.84	67	0.96		
20	58	0.79	62	0.88	69	0.97		
21	60	0.84	64	0.92				
22	61	0.86	65	0.93				
23	63	0.90	67	0.96				
24	64	0.92	69	0.97				
25	66	0.95	70	0.98				

Abbreviation: PR, percentile rank.

*t* value statistic (mean, 50; standard deviation, 10).

correlation coefficients between the HOST scales at the time of the first survey and 8 weeks later. All four HOST scales proved to be stable (Table 3). In the interview, the majority of parents declared that the HOST assessed all relevant topics and was easy to understand. Two parents suggested adding questions regarding the financial burden of families. Because this topic was considered of low importance for the assessment of sleep and daily troubles and the low number of parents suggesting inclusion of this topic, it was not added to the questionnaire.

### 3.6. Indices of distribution

We calculated reference values using data from our sample of 214 severely affected parents with children with SPMI. The respective *t* values and percentile ranks are presented in Table 7.

## 4. Discussion

Chronic pediatric diseases require continuous care in more than half of the affected children. Children with SPMI present an additional problem because nighttime care is mostly necessary. Parental psychologic well-being is markedly low and has a close relationship to the child's sleep in pediatric SPMI [6,10,11,31].

We aimed to develop an instrument to assess sleep and the daily troubles of parents with children with SPMI, which is called the HOST questionnaire. In a pilot study, the understandability, applicability, and economy of the tool were judged as adequate by parents. In the development of the HOST, an exploratory item analysis demonstrated that less informative items could be removed. The final version of the HOST consisted of 54 items. The HOST is meant to be used as a screening tool; it requires 15 min to be completed and is thus appropriate for this purpose. Its internal consistency (Cronbach  $\alpha$ , >0.7) and the test–retest reliabilities of its four scales ( $r_t$  > 0.7) confirm that the HOST is a helpful tool for the assessment of parental sleep-related burden in various life domains and over a long period of time. The normative values presented for the various

HOST scales (for *t* values, see Table 7) allow for an initial assessment of any impairments and clinical effects on the parents' well-being. The retest correlations of the HOST were predominantly high and highly significant. This finding supports our assumption that without any intervention parental burden and impairments remains unchanged, at least for a limited period of 8 weeks.

The convergent validity of the HOST is satisfactory and is reflected in its moderate to high positive correlation with established scales of the PSQI and the ESS. As expected, negative correlations between the HOST and parental SF-36 scores were observed. Higher perceived parental sleep-related burdens indicated worse parental health (e.g., somatic, psychic). Sleep-related impairments have the greatest effect on parental vitality, social functioning, and physical and emotional health. The scarcity of scientific data related to the lack of specific assessment tools for parents of children with SPMI hinder improvements of the clinical symptom-oriented medical and psychosocial care of the affected children and their parents.

The development of the HOST aimed to remedy this problem by facilitating the assessment of parental problems and the effects of having children with SPMI on everyday life. To the best of our knowledge, the HOST is the first tool explicitly developed for the assessment of sleep-related difficulties in this sample of parents. In contrast to the existing tools (SF-36, 30-item General Health Questionnaire [32]), the HOST considers the specific circumstances of affected families, including the necessity of nighttime care of older children, medical treatments, worries about the child's underlying disease, and disease-related fears of the future. The HOST is more specific than the PSQI. It assesses the stress factors in parent–child interactions such as child distress or poor parental sleep quality resulting from worries about the child; it also provides important information with direct implications for therapy. We believe that the HOST is useful for research purposes and in clinical practice as a screening tool for physicians and parents.

Some limitations of our study include its reliance on the parents' self-report to define their sleep problems and the lack of



corroboration of sleep symptoms with a more objective assessment, such as polysomnography or actigraphy. The response rate was 54% for the psychometric testing, which may have resulted in potentially skewed data. In our study 95% of the participants were mothers and 5% were fathers, and the mothers generally were the primary child caregivers. It is difficult to extrapolate information on the burden placed on fathers using our data. To further increase the validity of the HOST, use of an objective instrument that measures parental activity such as actigraphy or nighttime video recording would be helpful. For now the HOST consists of subjective parental ratings, which may demonstrate issues of convergent validity with objective measures. Further studies must be conducted to assess this relationship.

## 5. Conclusion

Ameliorating children's sleep difficulties and concomitantly reducing the sleep-related impairments of caregiving parents is essential in pediatric palliative care and other types of medical care. With the development of the HOST, we present a valid and reliable tool for the assessment of parents' 24-h sleep-related difficulties. The parental sleep-related burden is assessed according to the interests and needs of the severely disabled child in addition to the associated parental worries regarding the child and his or her development.

## Funding sources

No external funding supported our study.

## Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: <http://dx.doi.org/10.1016/j.sleep.2013.09.019>.

## Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.sleep.2013.09.019>.

## References

- [1] Cans C, Wilhelm L, Baille MF, Mazaubrun C, Grandjean H, Rumeau-Rouquette C. Aetiological findings and associated factors in children with severe mental retardation. *Dev Med Child Neurol* 1999;41:233–9.
- [2] Shevell MI, Ashwal S, Donley D, Flint J, Gingold M, Hirtz D, et al. Practice parameter: evaluation of the child with global developmental delay. *Neurology* 2003;60:367–80.
- [3] Contro N, Larson J, Scofield S, Sourkes B, Cohen H. Family perspectives on the quality of pediatric palliative care. *Arch Pediatr Adolesc Med* 2002;156:14–9.
- [4] Tietze A-L, Blankenburg M, Hechler T, Michel E, Koh M, Schlüter B, et al. Sleep disturbances in children with multiple disabilities. *Sleep Med Rev* 2011;16:117–27.
- [5] Blankenburg M, Tietze A-L, Hechler T, Hirschfeld G, Michel E, Koh M, et al. SNAKE: the development and validation of a questionnaire on sleep disturbances in children with severe psychomotor impairments. *Sleep Med* 2013;14:339–51.
- [6] Stores G, Wiggs L. Sleep disturbances in children and adolescents with disorders of development: its significance and management. London, England: Mac Keith Press; 2001.
- [7] Polimeni M, Richdale A, Francis A. The impact of children's sleep problems on the family and behavioural processes related to their development and maintenance. *EJAP* 2007;3:76–85.
- [8] Kandel I, Merrick J. The child with a disability: parental acceptance, management and coping. *Sci World J* 2007;7:1799–809.
- [9] Lam P, Hiscock H, Wake M. Outcomes of infant sleep problems: a longitudinal study of sleep, behavior, and maternal well-being. *Pediatr* 2003;111:e203–7.
- [10] Wolf LC, Noh S, Fisman SN, Speechley M. Brief report: psychological effects of parenting stress on parents of autistic children. *J Autism Dev Disord* 1989;19(1):157–66.
- [11] Meltzer LJ, Mindell JA. Impact of a child's chronic illness on maternal sleep and daytime functioning. *Arch Intern Med* 2006;166:1749–55.
- [12] Raina P, O'Donnell M, Rosenbaum P, Brehaut J, Walter SD, Russell D, et al. The health and well-being of caregivers of children with cerebral palsy. *Pediatr* 2005;115(6):e626–36.
- [13] Brehaut JC, Kohen DE, Raina P, Walter SD, Russell DJ, Swinton M, et al. The health of primary caregivers of children with cerebral palsy: how does it compare with that of other Canadian caregivers? *Pediatr* 2004;114:e182–91.
- [14] Hemmingsson H, Stenhammar AM, Paulsson K. Sleep problems and the need for parental night-time attention in children with physical disabilities. *Child Care Health Dev* 2009;35:89–95.
- [15] Wayte S, McCaughey E, Holley S, Annaz D, Hill CM. Sleep problems in children with cerebral palsy and their relationship with maternal sleep and depression. *Acta Paediatr* 2012;101:618–23.
- [16] Eisenhower AS, Baker BL, Blacher J. Preschool children with intellectual disability: syndrome specificity, behaviour problems, and maternal well-being. *J Intellect Disabil Res* 2005;49:657–71.
- [17] Quine L. Sleep problems in children with mental handicap. *J Intellect Disabil Res* 1991;35:269–90.
- [18] Colville GA, Watters JP, Yule W, Ax M. Sleep problems in children with sanfilippo syndrome. *Dev Med Child Neurol* 1996;38:538–44.
- [19] Boergers J, Hart C, Owens JA, Streisand R, Spirito A. Child sleep disorders: associations with parental sleep duration and daytime sleepiness. *J Fam Psychol* 2007;21:88–94.
- [20] Bruni O, Ottaviano S, Guidetti V, Romoli V, Innocenzi M, Cortesi F, et al. The sleep disturbance scale for children (SDSC): construction and validation of an instrument to evaluate sleep disturbances in childhood and adolescence. *J Sleep Res* 1996;5:251–61.
- [21] Malhotra NK, Birks DF. Marketing research: an applied orientation. 2nd ed. Boston (MA): Pearson; 2003.
- [22] Gentili A, Weiner DK, Kuchibhatla M, Edinger JD. Test-retest reliability of the Pittsburgh Sleep Quality Index in nursing home residents. *J Am Geriatr Soc* 1995;43:1317–8.
- [23] Buysse DJ, Reynolds 3rd CF, Monk TH, Berman SR, Kupfer DJ. The Pittsburgh Sleep Quality Index: a new instrument for psychiatric practice and research. *Psychiatr Res* 1989;28:193–213.
- [24] Carpenter JS, Andrykowski MA. Psychometric evaluation of the Pittsburgh Sleep Quality Index. *J Psychosom Res* 1998;45:5–13.
- [25] Doi Y, Minowa M, Uchiyama M, Okawa M, Kim K, Shibui K, et al. Psychometric assessment of subjective sleep quality using the Japanese version of the Pittsburgh Sleep Quality Index (PSQI-J) in psychiatric disordered and control subjects. *Psychiatr Res* 2000;97:165–72.
- [26] Backhaus J, Junghanns K, Broocks A, Riemann D, Hohagen F. Test-retest reliability and validity of the Pittsburgh Sleep Quality Index in primary insomnia. *J Psychosom Res* 2002;53:737–40.
- [27] Bullinger M, Ravens-Sieberer R. Stand der Forschung zur gesundheitsbezogenen Lebensqualität von Kindern: Eine Literaturanalyse. *Prävention und Rehabilitation* 1995;7:106–21.
- [28] Johns MW. Reliability and factor analysis of the Epworth Sleepiness Scale. *Sleep* 1992;15:376–81.
- [29] Lienert GA, Raatz U. Testaufbau und Testanalyse. 6th ed. Weinheim: Beltz Psychologie Verlags Union; 1998.
- [30] Fisseni H-J. Lehrbuch der psychologischen Diagnostik. 3rd ed. Hogrefe; 2004.
- [31] Hauenstein EJ. The experience of distress in parents of chronically ill children: potential or likely outcome? *J Clin Child Psychol* 1990;19:356–64.
- [32] Stevenson J, Simpson J, Bailey V. The factor structure of the GHQ-30 for mothers with young children. *J Reprod Infant Psychol* 1989;7:39–46.